

## Inflammatory Pseudotumor of the Liver: Report of a Case and Review of the Literature

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**Abstract:** Only 56 cases of inflammatory pseudotumor of the liver have been reported in the world literature since its first documentation in 1953. We report herein the case of a 68-year-old man incidentally found to have a lesion in the right lobe of the liver which closely resembled a neoplasm on imaging studies. Thus, partial hepatic resection was performed and histological examination of the resected specimen revealed a diagnosis of inflammatory pseudotumor. Surgical resection is the preferred treatment for inflammatory pseudotumor of the liver, especially in patients for whom a definite histologic diagnosis cannot be made preoperatively or by intraoperative frozen sections. In fact, most of the patients reported in the literature recovered uneventfully after local resection without any postoperative complications, as did our patient.

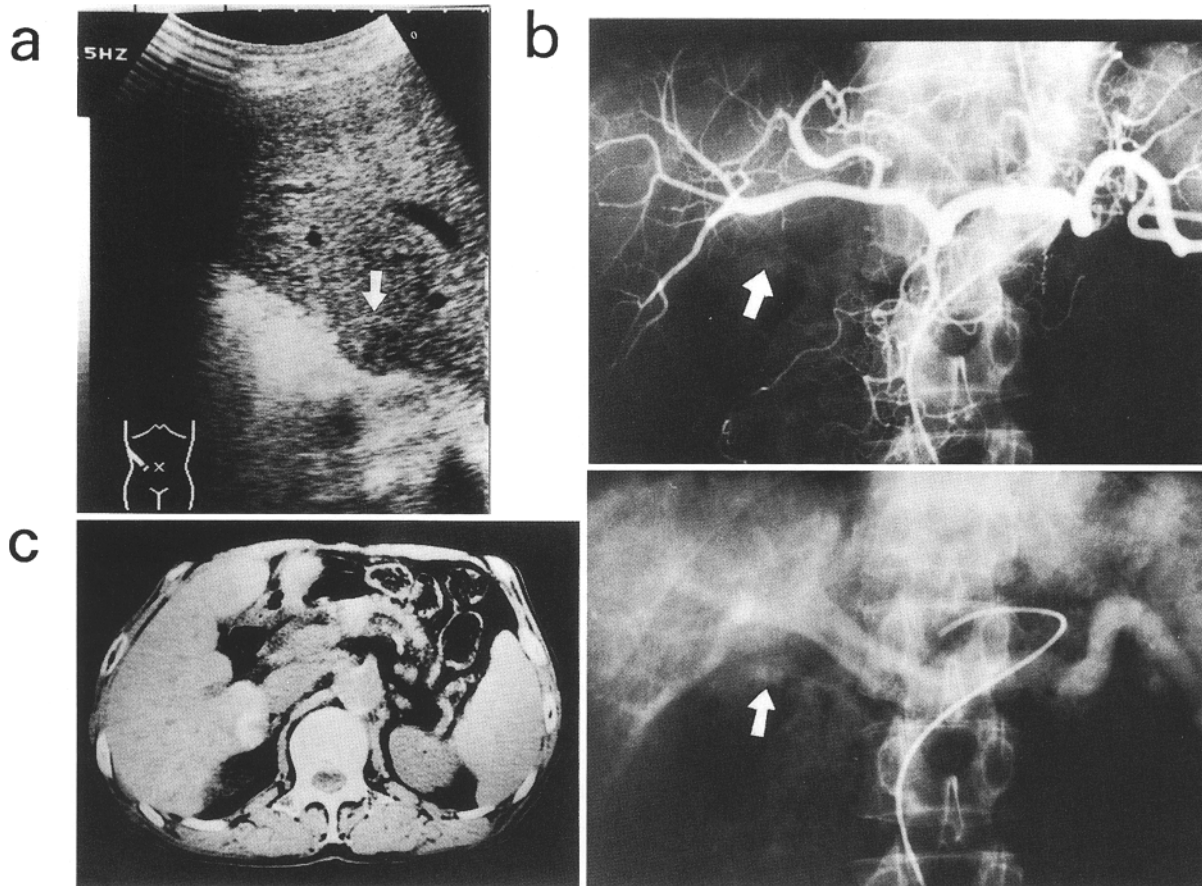
**Key Words:** plasma cell granuloma, liver mass, partial hepatectomy

### Introduction

Inflammatory pseudotumors of the liver simulating malignant neoplasms have recently been detected with advances in imaging diagnostic techniques.<sup>1-5</sup> We describe herein our experience of a patient with a nodular hepatic lesion which presented considerable diagnostic problems, especially in distinguishing it from hepatocellular carcinoma. However, it was eventually confirmed histologically to be a rare solitary inflammatory pseudotumor. Following this case report, we present our review of the literature and compare our case with those previously reported.

### Case Report

A 68-year-old male was admitted to our department for further investigation of a liver tumor that had been discovered by ultrasonography, after he presented to our clinic with a 2-week history of general fatigue and a 10-kg weight loss. He had not experienced any nausea, vomiting, fever, or antecedent flu-like syndrome, nor had he suffered from any previous liver or biliary tract disease. However, a history revealed that he had undergone partial gastrectomy for a gastric ulcer at the age of 56, and also that he had been diagnosed as having diabetes mellitus 1 year earlier. Physical examination on admission revealed that he was basically healthy, and no evidence of hepatosplenomegaly was found. A full blood count showed a hemoglobin of 12.8 g/dl, and a white cell count of 3800/l with 33% polymorphs, 44% lymphocytes, 12% monocytes, and 2% eosinophils. Other laboratory results revealed aspartate aminotransferase and alanine aminotransferase levels of 127 U/l and 67 U/l (the normal ranges being 11–35 and 3–35, respectively) and alkaline phosphatase and gamma-glutamyl transpeptidase levels of 408 U/l and 205 U/l (the normal ranges being 87–254 and 6–46, respectively). His serum was negative for hepatitis B surface antigen and hepatitis B surface antibodies, but positive for hepatitis C antibodies. The alpha-fetoprotein and carcinoembryonic antigen levels were normal. Ultrasonography of the abdomen revealed normal biliary ducts and no gallstones but a hypoechoic area indicating the presence of a liver tumor (Fig. 1a). A computed tomography (CT) scan confirmed the presence of a hypodense lesion in the posterior segment, and a lipiodol CT showed the accumulation of lipiodol in the lesion (Fig. 1c). Hepatic angiography revealed that the lesion was of a hypervascular nature in the arterial phase and that it appeared as a tumor stain in the venous phase (Fig. 1b). Under the preoperative diagnosis of hepatocellular carcinoma, partial hepatic



**Fig. 1.** **a** Abdominal ultrasonography revealed a hypoechoic mass in the posterior segment of the right hepatic lobe (*arrow*). **b** Hepatic angiography showed a hypervascular nature of the S6 lesion in the arterial phase (*top, arrow*) and a tumor

stain in the venous phase (*bottom, arrow*). **c** Lipiodol computed tomography (CT) showed the accumulation of lipiodol in the S6 lesion

resection of the posterior segment of the right hepatic lobe was performed. The cut surface of the resected material showed a well-defined, yellowish mass lesion, 19mm at its greatest axis (Fig. 2a). Histologically, the mass was predominantly composed of foamy histiocytes, as well as lymphocytes and plasma cells (Fig. 2b). Immunohistochemically, the plasma cells in the lesion showed positive staining for kappa and lambda light chains, revealing a polyclonal nature of the plasma cells. The hepatic tissue outside the lesion was essentially normal. Thus, a pathologic diagnosis of hepatic plasma cell granuloma compatible with a so-called inflammatory pseudotumor was made. The patient had an uneventful recovery and is currently well, 1 year after his operation.

### Discussion

Inflammatory pseudotumors of the liver may be defined as a group of diseases rather than a single disease entity. In fact, Someren<sup>6</sup> histologically classified inflam-

matory pseudotumors into three groups, namely (1) xanthogranuloma-type pseudotumors, (2) plasma cell granuloma-type pseudotumors, and (3) sclerosing pseudotumors. Their exact pathogenesis is still unclear; however, based on the clinical features and biologic behavior of the previously reported cases, it is generally regarded as a benign reactive inflammatory condition.<sup>7,8</sup>

To our knowledge, 57 cases of inflammatory pseudotumor of the liver, including that of our patient, have been reported in the world literature (Table 1). According to our review, there were 38 males and 19 females, with a male-to-female ratio of 2.0, ranging in age from 10 months to 83 years, with a mean age of 40.0 years. The lesion was located in the right lobe of the liver in 28 patients, in the left lobe in 15, and in the hepatic hilum in 4, and the tumor size ranged from 0.8 to 2.5 cm in diameter. Clinically, most patients with inflammatory pseudotumor have presented with fever, weight loss, and malaise, and investigations tended to reveal an elevated white cell count and erythrocyte sedimentation rate. Based on the symptomatology and laboratory

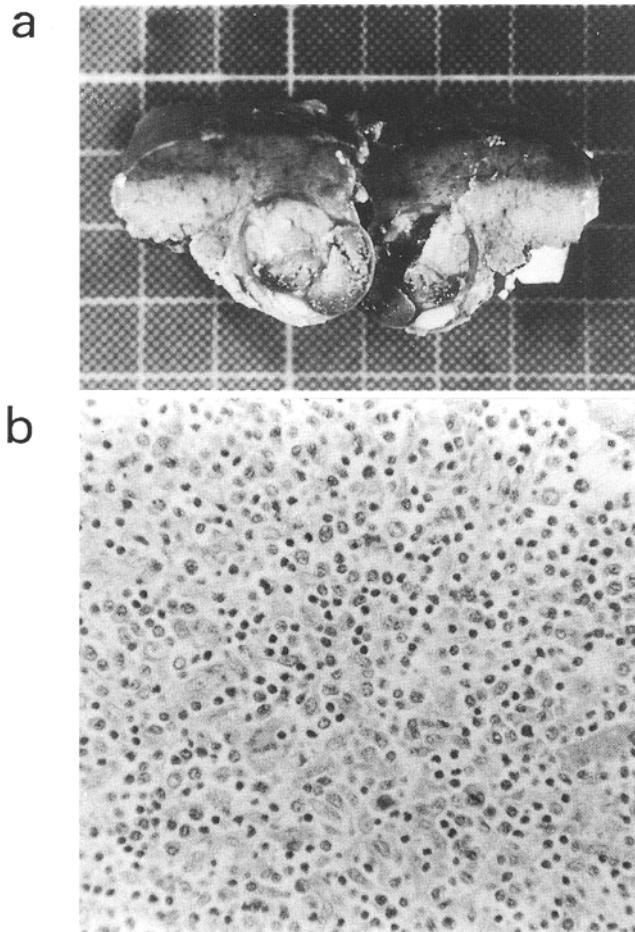
**Table 1.** Summary of reported cases of inflammatory pseudotumor of the liver

Reference	Age (years)	Sex	Race or nationality	Presentation	Solitary/multiple	Site	Size	Treatment	Outcome
Pack and Baker, 1953	40	M	German	Fever, weight loss, and right upper abdominal mass	Solitary	Right lobe	25 × 13 cm	Right lobectomy	Recovery
Haith et al., 1969	6	M	Caucasian	Fever, vomiting, jaundice, pruritus, and weight loss	Solitary	Porta hepatis	3 cm	Pancreatico-duodenectomy	Malabsorption
Hertzer et al., 1971	1	F	Caucasian	Fever, jaundice, and hepatomegaly	Solitary	Porta hepatis	?	Biopsy	Portal hypertension
Someren, <sup>6</sup> 1978	4.5	M	Caucasian	Fever and malaise	Solitary	Right lobe	8 × 6 × 6 cm	Right lobectomy	Recovery
Shinada et al., 1980	7.7	M	Japanese	Fever, abdominal pain, diarrhea, nausea, and vomiting	Multiple	Right lobe	4 × 4 = 3.5 cm and 1 × 1 cm	Right lobectomy	Recovery
Tanino et al., 1981	75	M	Japanese	General fatigue, appetite loss, and fever	Multiple	Right lobe	3.4 cm and 2.0 cm	Autopsy	Death
Higuchi et al., 1982	59	F	Japanese	A several-year history of fever and general fatigue	Solitary	Left lobe	4 cm	Left lobectomy	Recovery
Paineau et al., 1983	46	M	Unclear	Fever and weight loss	Solitary	Right lobe	7 cm	Right lobectomy	Recovery
Chen, 1984	29	M	Caucasian	Fever, weight loss, and hepatic mass	Solitary	Right lobe	6 cm	Wedge resection	Recovery
Heneghan et al., 1984	8	F	Unclear	Anorexia, abdominal pain, and jaundice	Multiple	Right lobe	?	Total hepatectomy and transplantation	Recovery
Kaneko et al., <sup>7</sup> 1984	10 months	F	Japanese	Vomiting and anemia	Solitary	Left lobe	4 cm	Left lobectomy	Recovery
Sasagawa et al., 1985	68	M	Japanese	Upper abdominal pain, fever, and weight loss	Solitary	Left lobe	10 cm	Left lobectomy	Recovery
Yamauchi et al., <sup>8</sup> 1985	47	M	Japanese	Fever and general fatigue	Solitary	Right lobe	10 × 8 cm	Biopsy	Recovery
Anthony and Telesinghe, <sup>10</sup> 1986	10	F	Unclear	Headaches, flushes, abdominal pain, diarrhea, and vomiting	Solitary	Right lobe	9 cm	Right lobectomy	Recovery
	16	M	Unclear	Fever	Multiple	Both lobes	Up to 3 cm	Biopsy	Recovery
	Child	M	Indian	Abdominal pain and night sweats	Solitary	Right lobes	5 cm	Biopsy	Recovery
	44	M	West Indian	A 7-year history of intermitten jaundice	Multiple	Both lobes	Up to 2 cm	Cholechojejunostomy and biopsy	Recovery
	57	M	Unclear	Nausea, vomiting, and upper abdominal pain	Multiple	Both lobes	Up to 2 cm	Biopsy, wedge biopsy of ampullae of Vater, and cholecystectomy	Recovery
Ushijima et al., 1987	48	M	Japanese	Fever and abdominal pain	Solitary	Right lobe	2.3 × 2.2 cm	Right lobectomy	Recovery
Grouls, <sup>11</sup> 1987	85	F	?	Unclear	Multiple	Right lobe	1.4 × 0.8 × 1.0 cm 0.7 × 0.8 × 0.7 cm	Autopsy	Death
Morita et al., <sup>12</sup> 1987	6.5	F	Japanese	Fever, abdominal pain, and vomiting	Solitary	Left lobe	6 × 9 cm		
Kessler et al., 1988	17	M	Arabian	Fever and abdominal pain	Solitary	Right lobe	8 cm	Laparotomy and wedge biopsy	Recovery
Joh et al., <sup>9</sup> 1988	58	F	Japanese	A several-year history of fever, abdominal pain, headache, and general fatigue	Solitary	Left lobe	1.7 × 1.5 × 1.4 cm	Left lateral segmentectomy	Recovery
Levitt et al., 1988	31	F	Caucasian	Fever and weight loss	Solitary	Right lobe	5 cm	Right lobectomy	Recovery

Table 1. Continued

Reference	Age (years)	Sex	Race or nationality	Presentation	Solitary/multiple	Site	Size	Treatment	Outcome
Li et al., <sup>1</sup> 1989	45	M	Chinese	Fever and abdominal pain	Solitary	Right lobe	8 × 7 cm	Right lobectomy	
	16	F	Chinese	Mass in upper abdomen	Solitary	Left lobe	6.5 × 4 cm	Life lobectomy	
	23	M	Chinese	Upper abdominal pain	Solitary	Right lobe	4 × 4 cm	Right lobectomy	
	52	F	Chinese	Upper abdominal pain	Solitary	Left lobe	2.5 × 2 cm	Left lobectomy	
	48	M	Chinese	Upper abdominal pain	Solitary	Left lobe	5 × 4.5 cm	Left lobectomy	
Lupovitch et al., 1989	54	M	Caucasian	Fever and weight loss	Multiple	Both lobes	Up to 6 cm	Antibiotics	Recovery
Standiford et al., 1989	77	M	Caucasian	Fever and weight loss	Solitary	Left lobe	4 × 6 cm	Wedge excision	Recovery
Horiuchi et al., <sup>2</sup> 1990	57	M	Japanese	Abdominal pain	Solitary	Left lobe	5 cm	Left lobectomy	Recovery
	27	M	Japanese	Headaches, diarrhea, arthralgia, and body weight loss	Solitary	Left lobe	5 cm	Left lobectomy	Recovery
	37	M	Japanese	Fever, abdominal pain, and jaundice	Solitary	Right lobe	6 × 4 × 4 cm	Autopsy	Death
	60	M	Japanese	General fatigue, and appetite loss	Solitary	Right lobe	2.7 cm	Partial hepatectomy	Recovery
	43		Japanese	Fever and abdominal pain	Solitary	Right lobe	3.0 cm	Right lobectomy	Recovery
	63	M	Japanese	Fever and abdominal pain	Solitary	Right lobe	4 × 4 × 4 cm	Autopsy	Death
	22	M	Japanese	No precise clinical data	Solitary	Right lobe	3 cm	Right lobectomy	Recovery
	83	F	Japanese	Fever	Solitary	Left lobe	4.7 × 4.2 cm	Biopsy	Recovery
	38	M	Japanese	Fever, abdominal distention, and pain	Solitary	Both lobes	Up to 3 cm	Laparotomy	Recovery
Andreola et al., <sup>3</sup> 1990	22	F	Caucasian	Abdominal pain	Solitary	Right lobe	13 × 12 × 9.5 cm	Right lobectomy	Recovery
	49	F	Caucasian	Abdominal pain	Multiple	Both lobes	?	Conservative	Death
Ikeda et al., 1990	43	M	Japanese	Fever and weight loss	Solitary	Porta hepatitis		Resection	Regression
Tsao et al., 1990	36	F	Chinese	?	?	?	?	?	?
Isobe et al., 1991	57	M	Japanese	Fever and abdominal pain	Solitary	Left lobe	?	Biopsy	Recovery
Jackson et al., 1991	67	M	Caucasian	Abdominal pain and weight loss	Multiple	?	?	Right lobectomy	Recovery
Pokorny et al., 1991	43	M	Caucasian	Jaundice	Solitary	Porta hepatitis		Biliary stenting	Well with tumor
Gollapudi et al., 1992	56	M	Caucasian	Fever and abdominal pain	Solitary	Right lobe	7 cm	Conservative	Recovery
Lopez et al., 1992	62	M	Caucasian	?	?	?	?	Spinicterotomy	Regression
Newbould et al., 1992	3	M	Caucasian	Incidental finding	Solitary	Left lobe	2 cm	Resection	Recovery
Shek et al., <sup>4</sup> 1993	31	F	Chinese	Fever and abdominal pain	Solitary	Right lobe	15 × 11 × 10 cm	Right lobectomy	Recovery
	35	F	Chinese	Fever, weight loss, and abdominal pain	Solitary	Right lobe	20 × 15 × 15 cm	Right lobectomy	Recovery
	33	M	Chinese	Incidental finding at laparotomy	Solitary	Left lobe	15 × 15 cm	Wedge biopsy	Recovery
	21	?	?	?	Solitary	?	1 cm	Excision	?
Nakajima et al., <sup>5</sup> 1993	67	F	Japanese	Abdominal pain and general malaise	Solitary	Right lobe	3.5 cm	Right lobectomy	Recovery
	46	F	Japanese	Abdominal pain	Solitary	Right lobe	2.7 × 2.5 × 2.3 cm	Right posterior segmentectomy	Recovery
Present case	68	M	Japanese	General fatigue and weight loss	Solitary	Right lobe	1.9 × 1.5 × 1.3 cm	Partial hepatic resection	Recovery

Adapted from Shek et al.,<sup>4</sup> except Joh et al.,<sup>9</sup> Grouls,<sup>11</sup> and Morita et al.<sup>12</sup>



**Fig. 2.** **a** Surgical specimen. The tumor measured  $1.9 \times 1.5 \times 1.3$  cm and the cut surface revealed a well-defined, yellowish mass lesion. **b** Histological findings. The lesion consisted of histiocytes, lymphocytes, and plasmacytes (H&E,  $\times 400$ )

findings, an underlying infective cause for inflammatory pseudotumor is likely.

Of clinical importance is differentially diagnosing between inflammatory pseudotumors and malignant neoplasms. Although most hepatocellular carcinomas reveal hypervascularity by angiography, a few demonstrate a hypovascular or avascular nature.<sup>9</sup> In addition to the angiography findings, the low density and good circumscription shown by CT and the mosaic pattern revealed by ultrasonography are frequently observed in inflammatory pseudotumors. Nevertheless, a space-occupying lesion expressed by the imaging findings may sometimes lead to an erroneous diagnosis.<sup>10</sup> In our patient, the imaging findings and histologic results of the intraoperative frozen section investigations failed to reveal the underlying nature of the lesion, and a definite diagnosis was only able to be made by performing histologic examination of the resected specimen. The prognosis of inflammatory pseudotumor of the liver is

generally considered to be good. In fact, none of the patients whose cases were reported in the literature died as a direct result of the tumor and most recovered after local resection without developing any postoperative complications. According to our review of the 57 cases, surgical resection was only performed when a preoperative diagnosis of hepatocellular carcinoma had been made. On the other hand, when the tumor cannot be removed surgically, steroid therapy has proven successful, as demonstrated by two reports of patients who were effectively treated by steroids after being deemed as having inoperable tumors.<sup>10</sup> Nevertheless, as it is difficult to differentiate inflammatory pseudotumors of the liver from hepatocellular carcinoma by imaging techniques, a needle biopsy of the tumor should always be performed. When inflammatory pseudotumor is able to be definitively diagnosed by the needle biopsy, conservative therapy is indicated; however, when a firm histologic diagnosis cannot be established preoperatively or by intraoperative frozen sections, surgical resection should be performed.

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